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OBSTRUCTIVE SLEEP APNOEA AFTER CLEFT PALATE SURGERY: A COMPREHENSIVE LITERATURE REVIEW

Surgery

Dr. Ritesh Kumar Singh* Postgraduate, Department of Oral and Maxillofacial Surgery, Manipal College of Dental Sciences, Mangalore. *Corresponding Author

Dr. Arvind Ramanathan Associate Professor, Department of Oral and Maxillofacial Surgery, Manipal College of Dental Sciences, Mangalore.

ABSTRACT

Obstructive Sleep Apnea (OSA) is a condition characterized by repeated episodes of partial or complete airway obstruction during sleep, leading to disrupted sleep patterns and systemic complications. The prevalence of OSA is significantly higher in individuals with craniofacial abnormalities, particularly those who have undergone cleft palate repair. Surgical correction of cleft palate is essential for improving speech, feeding, and aesthetic outcomes, but it may also result in postoperative airway alterations that increase the risk of OSA. This literature review aims to explore the prevalence, risk factors, diagnostic modalities, and management strategies for OSA following cleft palate surgery. Through a comprehensive evaluation of the available literature, including recent studies such as Ho et al.'s survey in Hong Kong, this article synthesizes findings on the anatomical, surgical, and functional factors contributing to OSA. Additionally, the review highlights the importance of early diagnosis and a multidisciplinary approach to managing OSA in this patient population. The review concludes by identifying gaps in current research and proposing future directions to optimize clinical outcomes in cleft palate patients at risk of developing OSA.

KEYWORDS

obstructive sleep apnoea, velopharyngeal insufficiency, cleft palate surgery, craniofacial surgery

INTRODUCTION

Cleft palate is a prevalent congenital craniofacial anomaly that manifests during embryonic development, typically within the first trimester of pregnancy. Affecting approximately 1 in 700 live births globally, it is characterized by a failure in the fusion of the palatal shelves, leading to an opening or gap in the roof of the mouth. (Brouillette et al., n.d.)The incidence of cleft palate varies across different geographical regions and ethnic backgrounds, with higher rates reported in certain populations such as Native Americans and Asians compared to lower prevalence in African and European populations. Cleft palate can occur in isolation or in conjunction with cleft lip, and in many cases, it is associated with various syndromic conditions, such as Pierre Robin sequence, Treacher Collins syndrome, or velocardiofacial syndrome. (Brouillette et al., n.d.)Regardless of its etiology, cleft palate presents significant challenges in early childhood, affecting essential functions such as feeding, speech development, hearing, and overall craniofacial growth.

The primary goal of cleft palate repair, or palatoplasty, is to restore functional anatomy by closing the defect and ensuring normal speech development and adequate oral function. Surgical correction is typically performed within the first year of life, although the timing of surgery can vary based on individual patient needs, local surgical protocols, and the presence of other associated anomalies. The success of cleft palate repair is evaluated based on several factors, including the restoration of velopharyngeal function, improved speech outcomes, and the patient's ability to feed and thrive without significant regurgitation or nasal escape. (Cielo et al., 2016)In some cases, additional secondary surgeries may be required to address residual speech issues or velopharyngeal insufficiency, further altering the postoperative anatomy.

However, while cleft palate surgery generally improves the functional and aesthetic outcomes for patients, it is not without its complications. One of the most notable and increasingly recognized postoperative concerns is Obstructive Sleep Apnea (OSA). OSA is a disorder characterized by repetitive episodes of partial or complete obstruction of the upper airway during sleep, leading to intermittent cessation of breathing (apnea) or reduced airflow (hypopnea). (Gorucu-Coskuner et al., 2020)These episodes result in hypoxemia, hypercapnia, and sleep fragmentation, ultimately contributing to a range of systemic complications. Left untreated, OSA can lead to significant morbidity, including cardiovascular issues such as hypertension, arrhythmias, and even heart failure, as well as neurocognitive impairments, daytime sleepiness, and metabolic disturbances like insulin resistance and obesity.(Silvestre et al., 2014)

In the context of cleft palate patients, the prevalence of OSA is notably higher compared to the general pediatric population. Numerous studies have demonstrated that children and adolescents who have undergone cleft palate surgery are at increased risk of developing OSA due to both anatomical and functional changes that occur following surgical repair. According to a study conducted by Ho et al. (2023), the prevalence of OSA in children with cleft lip and palate in Hong Kong was reported to be approximately 36%, a significantly higher rate than the estimated 1% to 4% prevalence of OSA in the general pediatric population . This discrepancy highlights the importance of recognizing and addressing the unique airway challenges faced by patients with cleft palate.

The pathophysiology of OSA in this patient population is multifactorial and often involves a combination of anatomical abnormalities, such as altered maxillary and mandibular development, scarring from surgical interventions, and residual velopharyngeal insufficiency. The upper airway in cleft palate patients is often structurally compromised due to the congenital defect and subsequent reconstructive efforts, which may lead to a narrowed airway or areas of obstruction, particularly during sleep when muscle tone is reduced.(Deepa & Ramnarayan, 2013) Additionally, secondary surgeries, such as pharyngeal flap procedures used to address velopharyngeal insufficiency, can further contribute to upper airway obstruction by restricting airflow through the nasal and pharyngeal passages.

Velopharyngeal insufficiency, a common complication of cleft palate repair, refers to the inability of the soft palate to close properly against the posterior pharyngeal wall, which can result in speech abnormalities and nasal regurgitation. While surgical techniques aim to correct this insufficiency, they can also disrupt the delicate balance of the upper airway, predisposing patients to obstructive events during sleep. For example, the use of pharyngeal flap surgery, which involves creating a flap of tissue to close the velopharyngeal gap, can obstruct airflow through the nasopharynx, particularly in patients with small oropharyngeal airways.(Bs et al., 2014)

Moreover, cleft palate patients may also exhibit abnormal craniofacial growth patterns, particularly in the maxilla and mandible. These anomalies, such as maxillary hypoplasia or retrognathia (a recessed lower jaw), can reduce the space available for the tongue and other soft tissues of the oropharynx, further exacerbating the risk of airway obstruction during sleep. In syndromic patients with craniofacial abnormalities like Pierre Robin sequence, which is characterized by a small jaw and a tendency for the tongue to fall backward into the airway, the risk of OSA is even more pronounced. (Jamieson et al., 2018)The confluence of these anatomical factors, combined with the impact of surgical interventions, creates a unique set of challenges in managing OSA in cleft palate patients.

The clinical presentation of OSA in cleft palate patients is often subtle and may be overlooked, as some of the hallmark symptoms of OSA, such as snoring, gasping for breath, or witnessed apneas, may not be as pronounced in this population. Additionally, speech and nasal abnormalities may mask or complicate the diagnosis of OSA, particularly in younger children. As a result, there is a growing recognition of the need for heightened vigilance among healthcare providers in screening for OSA in post-cleft palate surgery patients.(MacLean et al., 2009)

In recent years, advancements in diagnostic modalities, particularly the use of polysomnography (PSG), have improved the ability to accurately diagnose OSA in this population. PSG remains the gold standard for OSA diagnosis, offering a comprehensive assessment of sleep architecture, respiratory events, and oxygen saturation. Other diagnostic tools, such as cephalometric radiographs and nasopharyngoscopy, can provide valuable insights into the anatomical factors contributing to airway obstruction and can assist in preoperative planning for surgical interventions aimed at managing OSA.(Jungbauer et al., 2022)

Given the complexity of managing OSA in cleft palate patients, a multidisciplinary approach is essential. Collaboration between otolaryngologists, maxillofacial surgeons, sleep specialists, orthodontists, and speech therapists is often required to develop individualized treatment plans that address both the functional and anatomical aspects of OSA. Management strategies may include non-surgical interventions such as Continuous Positive Airway Pressure (CPAP) therapy or, in more severe cases, surgical options like maxillomandibular advancement or pharyngeal surgeries. (Abdel-Aziz, 2012)However, the risk of postoperative complications, including speech disturbances and velopharyngeal insufficiency, must be carefully considered in the decision-making process.

Aims And Objectives

The primary aim of this literature review is to provide a comprehensive overview of the current understanding of OSA in patients following cleft palate surgery. Specific objectives include:

1. To assess the prevalence of OSA in children and adolescents who have undergone cleft palate surgery.
2. To identify the anatomical, surgical, and systemic risk factors that contribute to the development of OSA in this population.
3. To review the various diagnostic methods used to identify OSA in cleft palate patients.
4. To evaluate the effectiveness of surgical and non-surgical management strategies for OSA.
5. To highlight the need for multidisciplinary approaches in managing OSA and propose future research directions.

MATERIALS AND METHODS

The literature search for this review was conducted across multiple electronic databases, including PubMed, Scopus, and Google Scholar. The search terms used included "Obstructive Sleep Apnea," "cleft palate," "craniofacial anomalies," "OSA diagnosis," "OSA management," and "post-surgical OSA." The review focused on studies published in the past 15 years to ensure that the most up-to-date research was included. A total of 20 articles were selected for this review based on their relevance to the topic, with a particular focus on studies involving pediatric populations with cleft palate who were diagnosed with OSA.

Inclusion criteria for the studies reviewed included clinical trials, observational studies, and case reports that specifically addressed OSA in post-cleft palate surgery patients. Studies that lacked clinical or polysomnographic data for OSA diagnosis were excluded, as were those that focused solely on adult populations. The primary outcomes of interest were the prevalence of OSA, risk factors associated with its development, and the effectiveness of different treatment modalities.

Prevalence of OSA in Post-Cleft Palate Surgery Patients

The prevalence of OSA in patients who have undergone cleft palate surgery has been extensively studied, with results indicating a significantly higher incidence of OSA in this population compared to the general pediatric population. Studies such as that conducted by Ho et al. in Hong Kong (2023) report a prevalence of OSA of approximately 36% in children and adolescents with a history of cleft lip and palate surgery. This elevated prevalence is consistent with other studies, which estimate that 30% to 40% of post-surgical cleft palate patients develop OSA.

Several factors contribute to this increased risk. The alteration of airway anatomy following cleft palate repair can predispose patients to upper airway obstruction, particularly during sleep when muscle tone decreases. The formation of scar tissue and the reconfiguration of the velopharyngeal area can further exacerbate airway patency issues, leading to recurrent episodes of airway collapse during sleep. In addition, some patients may experience residual velopharyngeal insufficiency or nasal obstruction after surgery, both of which are risk factors for OSA.

Interestingly, the prevalence of OSA in cleft palate patients appears to vary based on the type of surgical technique used for cleft repair, the age at which the surgery is performed, and the presence of comorbid conditions such as obesity or syndromic craniofacial anomalies. For instance, patients with syndromic conditions such as Pierre Robin sequence or Treacher Collins syndrome, which are often associated with cleft palate, have been shown to have an even higher prevalence of OSA due to their complex craniofacial structures.

Risk Factors For Developing Osa Post-cleft Palate Surgery

The risk factors for developing OSA following cleft palate surgery can be categorized into anatomical, surgical, and systemic factors. Anatomically, patients with cleft palate often have altered craniofacial morphology that predisposes them to airway obstruction. The cleft deformity itself can lead to abnormalities in the maxillary and mandibular structures, which may compromise the size and patency of the airway. Furthermore, the soft tissues of the palate and pharynx may be less mobile or scarred following surgical repair, contributing to airway obstruction during sleep.

Surgical factors also play a significant role in the development of OSA. The type of cleft repair technique used can influence the likelihood of OSA, with some techniques, such as pharyngeal flap surgery, potentially leading to postoperative airway obstruction. Additionally, patients who require secondary surgeries to address velopharyngeal insufficiency or speech abnormalities may be at greater risk for developing OSA due to further alterations in airway dynamics. (Liao, Samuel Noordhoff, et al., n.d.)The timing of surgery is another important consideration, as early surgical intervention may allow for better airway development and reduce the risk of OSA later in life.

Systemic risk factors, including obesity, are well-documented contributors to OSA in both the general population and in patients with cleft palate. Overweight and obese children are more likely to experience airway obstruction due to the accumulation of fatty tissue around the upper airway. Furthermore, children with syndromic craniofacial abnormalities often have additional risk factors for OSA, such as midface hypoplasia or glossoptosis, which can exacerbate airway obstruction during sleep.(Liao, Claudia Yun, et al., n.d.)

Diagnostic Approaches to OSA in Post-Cleft Palate Surgery Patients

The diagnosis of OSA in patients with a history of cleft palate surgery typically involves a combination of clinical assessment and objective testing. Polysomnography (PSG) remains the gold standard for diagnosing OSA, as it provides a comprehensive evaluation of sleep architecture, respiratory events, and oxygen saturation. PSG is particularly important in this patient population, as clinical symptoms of OSA, such as snoring or witnessed apneas, may be less apparent or masked by speech and nasal abnormalities.

In addition to PSG, several screening tools can be used to assess the likelihood of OSA in cleft palate patients. The Pediatric Sleep Questionnaire (PSQ) and the Epworth Sleepiness Scale (ESS) are commonly used to evaluate symptoms of sleep-disordered breathing and daytime sleepiness, respectively. However, these tools may not be as sensitive in detecting OSA in patients with craniofacial anomalies, as their symptoms may differ from those of the general population.(Liao, Ming-lung Chuang, et al., n.d.)

Radiographic evaluations, including cephalometric radiographs and nasopharyngoscopy, are also valuable in assessing the anatomical contributions to airway obstruction in post-surgical cleft palate patients. Cephalometric analysis allows for the measurement of craniofacial structures and their relationship to the airway, while nasopharyngoscopy provides direct visualization of the upper airway

during respiration. (Robison & Otteson, 2011)These imaging modalities can be particularly useful in pre-surgical planning and in identifying patients who may benefit from further surgical intervention to address OSA.

Management of OSA in Post-Cleft Palate Surgery Patients

The management of OSA in patients who have undergone cleft palate surgery requires a multidisciplinary approach that incorporates both surgical and non-surgical treatment modalities. Non-surgical management, particularly the use of Continuous Positive Airway Pressure (CPAP), is the first-line treatment for many patients with OSA. CPAP works by delivering a continuous stream of air to the upper airway, preventing its collapse during sleep. CPAP is highly effective in reducing apneic events and improving sleep quality, but patient compliance can be challenging, particularly in young children or those with developmental delays.(Cielo & Marcus, 2015)

For patients who do not tolerate CPAP or who have anatomical abnormalities that contribute significantly to airway obstruction, surgical management may be necessary. Adenotonsillectomy is a common surgical intervention for pediatric OSA, particularly in cases where hypertrophy of the adenoids and tonsils is a contributing factor. In cleft palate patients, however, the role of adenotonsillectomy is more complex, as it may exacerbate velopharyngeal insufficiency in some cases.

Other surgical options for managing OSA in cleft palate patients include maxillomandibular advancement (MMA) and distraction osteogenesis. (Ettinger et al., 2012)These procedures aim to expand the airway by repositioning the maxilla and mandible, thereby increasing the space available for airflow. Pharyngeal surgeries, such as uvulopalatopharyngoplasty (UPPP) or pharyngeal flap surgery, can also be performed to reduce airway obstruction. However, these procedures carry the risk of postoperative complications, such as velopharyngeal insufficiency or speech disturbances, and should be carefully considered in the context of each patient's unique anatomy and functional needs.

DISCUSSION

The relationship between cleft palate surgery and the development of OSA is complex and multifactorial. While cleft palate repair is essential for improving speech, feeding, and aesthetic outcomes, it can also result in anatomical changes that increase the risk of airway obstruction. The prevalence of OSA in this population is notably higher than in the general pediatric population, with studies reporting rates as high as 36%. Risk factors for OSA include anatomical alterations resulting from the cleft deformity and surgical repair, as well as systemic factors such as obesity and syndromic craniofacial anomalies.(Ho et al., 2023; Muntz et al., 2008)

The management of OSA in post-surgical cleft palate patients requires a multidisciplinary approach, incorporating both non-surgical and surgical interventions. CPAP remains the first-line treatment for most patients, but surgical options such as adenotonsillectomy, maxillomandibular advancement, and pharyngeal surgeries may be necessary in cases where anatomical abnormalities contribute significantly to airway obstruction. (Greenlee et al., 2019)Early diagnosis and intervention are crucial for preventing the long-term complications associated with untreated OSA, including cardiovascular, neurocognitive, and metabolic disorders.

Limitations Of Existing Literature

While significant progress has been made in understanding the relationship between cleft palate surgery and OSA, several limitations in the existing literature remain. Many studies are limited by small sample sizes, which may not provide a comprehensive representation of the population. Additionally, there is a lack of randomized controlled trials evaluating the effectiveness of different management strategies for OSA in cleft palate patients. Most studies are observational in nature, which limits the ability to draw definitive conclusions about causality.

Geographical and population differences also present a challenge in comparing study outcomes. Variations in surgical techniques, healthcare access, and follow-up care across countries may influence the reported prevalence of OSA and the effectiveness of different treatment modalities. Furthermore, there is a need for long-term follow-up studies to assess the progression or resolution of OSA in cleft palate patients over time.

CONCLUSION

In conclusion, OSA is a significant and under-recognized complication in patients who have undergone cleft palate surgery. The prevalence of OSA in this population is higher than in the general pediatric population, and its development is influenced by a combination of anatomical, surgical, and systemic factors. Early diagnosis and intervention are essential for preventing the long-term complications associated with untreated OSA. A multidisciplinary approach that includes both surgical and non-surgical management options is necessary to optimize outcomes for these patients. Future research should focus on conducting larger, randomized controlled trials and long-term follow-up studies to better understand the natural history of OSA in cleft palate patients and to evaluate the effectiveness of different treatment modalities.

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